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관골부에 발생한 결절성 근막염의 미용적 고찰

박태환 · 장충현

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Nodular Fasciitis of the Zygomatic Area: A Case Report and Its Aesthetic Considerations

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Nodular fasciitis is the most common benign mesenchymal tumor that can be mistaken both clinically and pathologically for soft tissue sarcoma due to its infiltrative growth pattern. It most commonly appears in the extremities and trunk. However, occurrence in the zygomatic area is extremely rare. We present a case of 28-year-old man, who was characterized with a firm, tender, clearly visible mass in his left zygomatic area. We performed a local excision, together with microscopic and immunohistochemical analysis. The mass was eventually diagnosed as nodular fasciitis in the zygomatic area. No postoperative oncological treatment was done and the postoperative follow-up was maintained up to the present time without any tumor recurrence. Nodular fasciitis should be considered in the differential diagnosis when a rapid growing subcutaneous mass is detected in this area. Accurate diagnosis, appropriate treatment and follow-up are mandatory to avoid overly aggressive treatment and limit treatment-related morbidities. Moreover, the disease itself and its surgical treatment may cause aesthetic compromise and the aesthetic implications have been discussed.

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Key Words: Fasciitis, Sarcoma, Aesthetic surgery

I. INTRODUCTION

Nodular fasciitis (NF) is an idiopathic, benign, rapid-growing, fibroblastic, and myofibroblastic proliferation that affects the subcutis and typically involves the upper extremities (48%), followed by the trunk (20%), head and neck (15~20%), and the lower extremities.[1] NF accounts for 0.18% of all pathological diagnosis; however, the exact incidence is unknown.[2]

Konwaler et al. first described and officially named "nodular fasciitis" in 1955. NF is also referred to as pseudosarcomatous fasciitis, pseudosarcomatous fibromatosis, proliferative fasciitis, and infiltrative fasciitis.[3] NF can be misdiagnosed as a malignant neoplasm, most commonly as a soft tissue sarcoma, both clinically and pathologically due to its malignant appearance, including its high cellularity, increased mitotic activity, and infiltrative growth pattern.[4]

In the literature, 10% to 15% of cases are associated with trauma. Although the lesion is considered a self-limiting reactive process associated with previous trauma, the specific etiology of nodular fasciitis is poorly understood.

The present case highlights some noteworthy features. First, only four other cases of NF of the zygomatic area have been

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reported.[3,5-7] Second, we present a detailed discussion of the diagnosis and treatment with reference to the pertinent literature.

II. CASE REPORT

A 28-year-old Korean man presented with a firm, tender, clearly visible mass in his left zygomatic area, that had enlarged rapidly to approximately 2.5 cm in diameter over the previous 4 months (Fig. 1). His family history was unremarkable and he had no history of infection or trauma. However, he suffered from Graves' disease with Graves' ophthalmopathy and was treated with methimazole. At the age of 26, the patient presented with signs and symptoms of hyperthyroidism, with suppressed TSH (0.01 uIU/mL), elevated free thyroxine (2.81 ng/dL), elevated TSH receptor antibody (5.41 IU/mL), elevated thyroglobulin antibody (4.93 IU/mL), and elevated microsomal antibody (72.53 IU/mL). The patient reported easy fatigue and intermittent diplopia. A physical examination revealed a firm, tender, and mobile nodular mass in his left zygomatic area and swollen thyroid gland with chemosis as well as injection of the conjunctiva, and limitation of EOM over than 30º in downgaze. Lagophthalmos was 1.5 mm in both eyes and exophthamos was 22 mm and 23 mm in the left and right eye, respectively. Computed tomography (CT) with contrast was performed 5-months before the visit to our department revealed increased retroorbital fat with slightly enlarged inferior rectus muscles consistent with Graves' ophthalmopathy (Fig. 2, Above). At that time, a well-defined, 0.7 cm sized, round shaped, well-enhancing intramuscular mass of the left zygomatic area was also noticed on a CT scan (Fig. 2, Below). However the patient was unaware of it, since the mass was not visible and palpated. Since then, the mass grew rapidly and the patient was referred to our department to exclude other soft tissue malignancies.

A local excision of the lesion infiltrating into the zygomaticus major muscle was performed with perilesional approach under local anesthesia and the defect was closed primarily with the muscle reapproximated.

The gross pathology of the specimens showed a grayish white well-demarcated, unencapsulated, firm, soft tissue mass measuring 2.6 × 1.6 × 1.5 cm in size (Fig. 3).

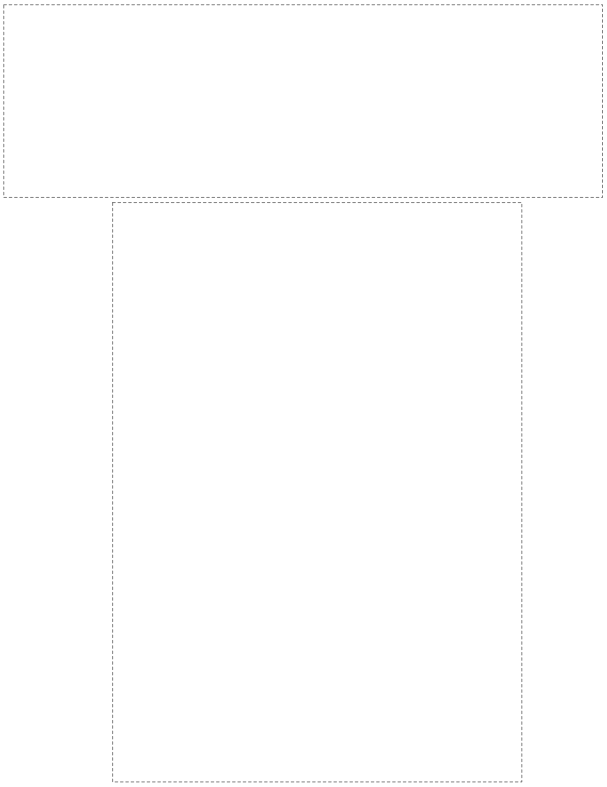

Fig. 1. Preoperative appearance of the patient. Approximately 2.5 cm sized, firm, tender, clearly visible mass in his left zygomatic area (Above) Frontal view (Below) Close up view.

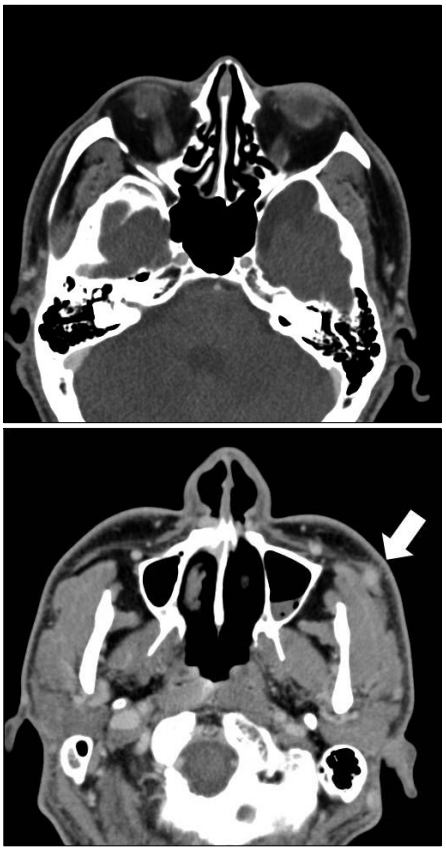

Fig. 2. Contrast-enhanced axial CT scan 5 months before presentation to our department. (Above) Increased retroorbital fat with slightly enlarged inferior rectus muscles consistent with Graves' ophthalmopathy. (Below) A well-defined, 0.7 cm sized, round shaped mass in the zygomaticus major muscle (white arrow).

The histological examination revealed alternating hypercellular and hypocellular areas, composed of uniform and plump spindle cell proliferation without atypia. Frequent mitotic figures were observed, but no atypical mitosis. Multifocal microhemorrhages were also noted (Fig. 4).

The immunohistochemical staining showed that the lesion stained positive for SMA, and CD68, and negative for cytokeratin, CD34, S-100 protein (Fig. 5). The mass was eventually diagnosed as NF.

The patient did not undergo any postoperative oncological treatment. The postoperative follow-up was maintained up to the present time, 2 years after excision, without any tumor recurrence.

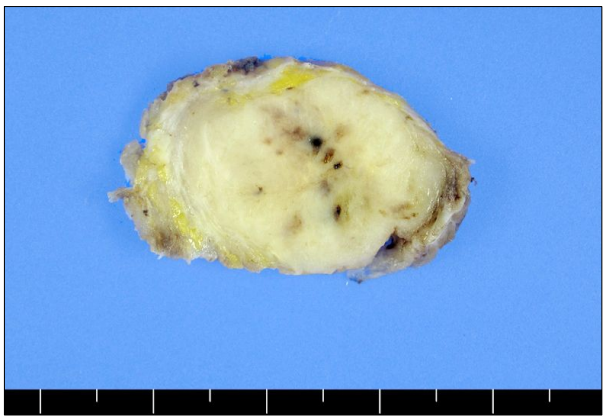

Fig. 3. Gross specimen. A grayish-white, well-demarcated, unencapsulated, firm, soft tissue mass measuring 2.6 × 1.6 × 1.5 cm in size.

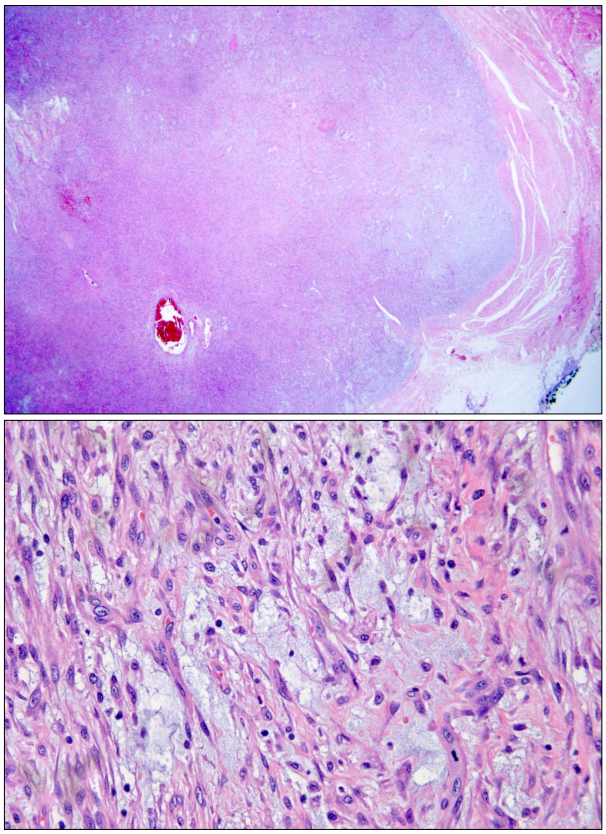

Fig. 4. Microscopic examination. (Above) Well-defined border with multifocal microhemorrhages (H&E stain, × 1.25). (Below) Alternating hypercellular and hypocellular areas, composed of uniform and plump spindle cell proliferation without atypia (H&E stain, × 400).

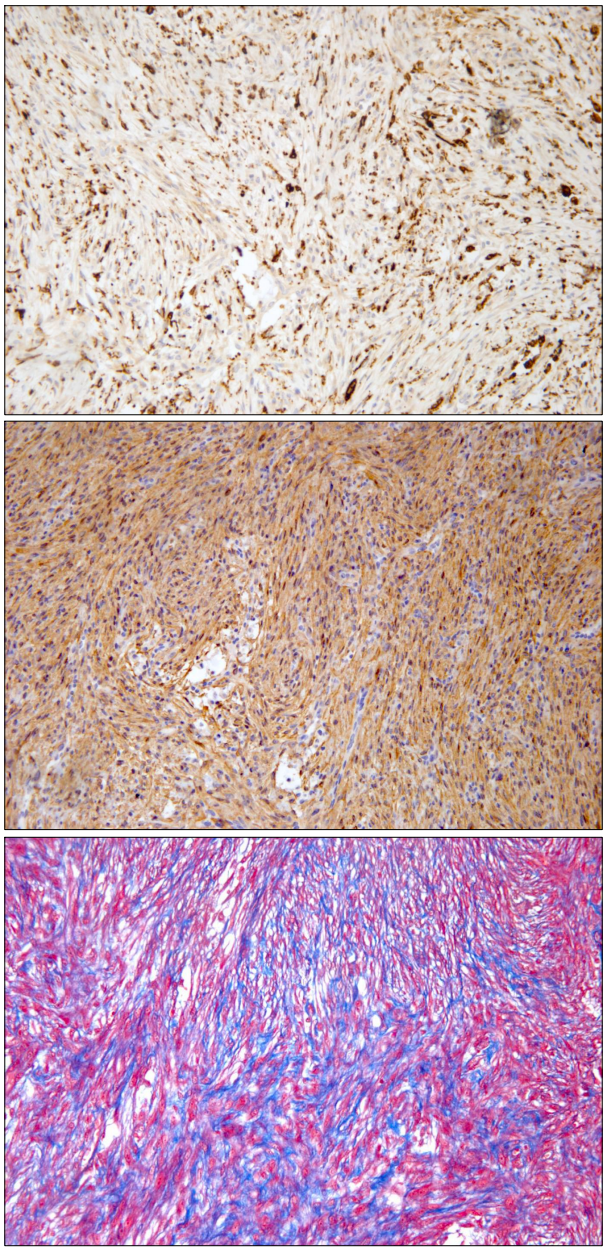

Fig. 5. Immunohistochemical examination. (Above) Tumor stained positive for CD68. (Center) Tumor stained positive for SMA. (Below) Masson-trichrome stain showing proliferation of collagen and myofibroblasts.

III. DISCUSSION

NF can appear at any age. However, it occurs most commonly in the third and fourth decades of life, with females being more frequently affected. Multiple lesions are extremely rare, and the size of the lesions can vary from 0.5 cm to 10 cm, with most being less than 4 cm in diameter.[1] Three subtypes of NF can be identified based on the location of the lesion, which may be in the subcutaneous, intramuscular, or fascial tissues. In particular, the intramuscular subtype, may mimic soft tissue malignancies during both the clinical and the imaging evaluation because of their larger size, deeper location, and less defined borders.

The most common clinical presentation is a solitary, rapidly growing mass that is frequently associated with pain and tenderness. Less frequently, perineural extension can cause numbness, paresthesia, and shooting pain. However, the clinical findings of NF appear to be nonspecific and do not alone support diagnosis.

Most cases of NF are well defined but non-encapsulated. Therefore, the margins appear irregular, and the lesion seems to infiltrate the surrounding tissue, masquerading as a malignant neoplasm. For this reason, several malignant diagnoses, including fibrosarcoma, leiomyosarcoma, and spindle cell carcinoma, should be included on the differential diagnosis. However, NF differs from malignancy in several distinct ways and is not a presarcoma or transformative lesion.

On ultrasonography, NF has been described as solid, hypoechoic, or mixed echoic, whereas on CT, the mass has been described as well defined, homogeneous, and of soft tissue density, even though moderate to strong enhancement has been reported. On magnetic resonance imaging, NF is described as having variable signal intensities that might indicate variability in cellularity and vascularity, even though high signal intensities on T1- and T2-weighted MR images are common.[8] Although imaging techniques might be useful in the diagnosis of NF, NF lesions can be difficult to distinguish from other soft tissue lesions, because most soft tissue masses have a rather nonspecific imaging appearance.

A tissue examination would probably be the diagnostic test of choice. The characteristic microscopic findings include haphazardly arranged pleomorphic spindle cells in a myxoid stroma with abundant mitoses without atypical figures and the stroma contains a mild, predominantly lymphocytic infiltrate with erythrocyte extravasion.[4] Its focal infiltration into surrounding tissues, its densely cellular nature, and the presence of mitotic figures may be concerning for malignancy. However, nodular fasciitis can be distinguished from a malignant process by the lack of cytologic atypia combined with the presence of focal myxoid areas and extravasated red blood cells. Immunohistochemistry has shown that the spindle cells in NF contain vimentin and sometimes actin, but do not contain desmin, keratin, or S-100 protein.

From an aesthetic view, nodular fasciitis itself causes tissue distortion, with rapid growing characteristics. In addition, scar may result from subcutaneous fat removal, since extraoral surgical approach is required. Surgeons inevitably are burdened with these aesthetic complications. However, the risk should always be considered and minimized with appropriate surgical technique. The reconstructive challenge in NF in the face comprises of large tissue defects which need cover if bone or vital structures are exposed. However, NF does not require a wide margin of resection, and does not compounds this since primary closure is not difficult to perform. In our patient, the treatment lay in being able to create a near-normal-looking left zygomatic area of the face with minimal scar formation.

Standard treatment consists of local excision with a minimum surgical margin and this usually results in permanent resolution. However, resection with a negative surgical margin is not mandatory because spontaneous regression without metastases and the recurrence of incompletely excised lesions often occurs.[2] In addition, NF may spontaneously regress. For this reason, a period of watchful waiting can be an alternative treatment in some cases with somewhat smaller lesions after initial biopsy demonstrating NF, considering its aesthetic aspects. Therefore, an awareness of the subtle distinctions between NF and soft tissue sarcoma is critical to avoid inappropriate measures and limit potential treatment-related morbidities.

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